CASE REPORT

Recurrent unicystic ameloblastoma in a child

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ABSTRACT

Unicystic ameloblastoma (UCA) is a clinical subtype of ameloblastoma that is considered prognostically different. The purpose of this report is to present a case of UCA showing dual radiographic pattern in a child. A detailed study of the lesion was carried out in an 8 year old female child who presented to our department of oral and maxillofacial pathology. Clinical, radiological and histopathological findings were recorded. In March 2005, a painless swelling in the left side of the mandible was noted, which on radiographic examination showed a unilocular radioluceny enclosing the crown of mandibular left permanent second molar, extending between the left first permanent molar and anterior margin of the ramus. Histopathologic diagnosis was UCA. The lesion was treated by enucleation. The patient returned with recurrence in 2009, at this time the lesion radiographically presented as a multilocular radiolucency with a soap bubble appearance, extending between the anterior border of the ramus and second premolar. Histopathologic diagnosis was UCA. The lesion was treated by segmental resection with immediate reconstruction. Although a number of treatment modalities are available to treat UCA, many factors need to be taken into consideration in the treatment of UCA in children.

Key words: Multilocular, second molar, unilocular, unicystic ameloblastoma

INTRODUCTION

Unicystic ameloblastoma (UCA) was first described by Robinson and Martinez,[1] to delineate its less aggressive course compared to solid ameloblastoma. With the exception of age of onset and biologic outcome,[1,2] this entity does not differ much from the solid ameloblastoma.[2] The purpose of this report is to present a case of UCA showing a pericoronal radiolucency in the primary lesion and multilocular radiolucency in the recurrent lesion.

CASE REPORT

An 8-year old girl presented in March 2005, with a painless swelling on the left side of the mandible of 1 month duration. Extra orally, slight asymmetry was noted and intra orally, the swelling was seen extending from mandibular left permanent first molar to the retro molar region. Panoramic radiograph showed a unilocular radiolucency between mandibular left first permanent molar and anterior margin of the ramus, enclosing the crown of permanent second molar [Figure 1]. Aspiration showed positive results. Clinical provisional diagnosis was dentigerous cyst.

Histologically, the incisional specimen was characterized by a cystic lining with ameloblast-like basal cells and stellate reticulum-like overlying cells. The fibrous wall was devoid of odontogenic islands and slightly myxoid in character [Figure 2]. In April 2005, the lesion was enucleated and a single cystic sac was noted. The post surgical specimen showed same features as incisional biopsy specimen but the fibrous wall showed inactive odontogenic islands [Figure 2-Inset]. The lesion was typed as simple unicystic ameloblastoma. She was lost to follow-up.

The patient returned with the complaints of pain and swelling in 2009. Extra orally, mild asymmetry was noted on the left side of the mandible. It was hard and tender to palpation. Intra orally, the swelling was found in the edentulous region in relation to missing second permanent molar. Radiographically (panoramic and lateral oblique view radiographs), a multiculocular, soap bubble, radiolucency was found between the anterior border of the ramus and second premolar

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DOI: 10.4103/0973-029X.84513

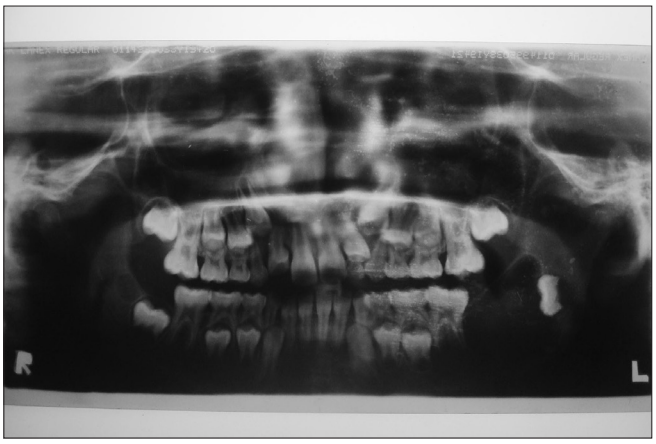

Figure 1: Panoramic radiographic examination of the initial lesion showing a unilocular radiolucency between mandibular left first permanent molar and anterior margin of the ramus, enclosing the crown of the permanent second molar

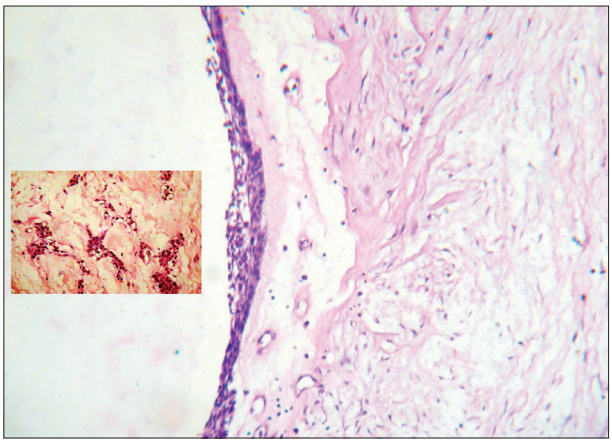

Figure 2: Histopathological examination of the incisional specimen characterized by a cystic lining ameloblast-like basal cells and stellate reticulum-like overlying cells. The fibrous wall is slightly myxoid in character. INSET – fibrous wall showing inactive odontogenic rests (H and E stain, 60×; 240×)

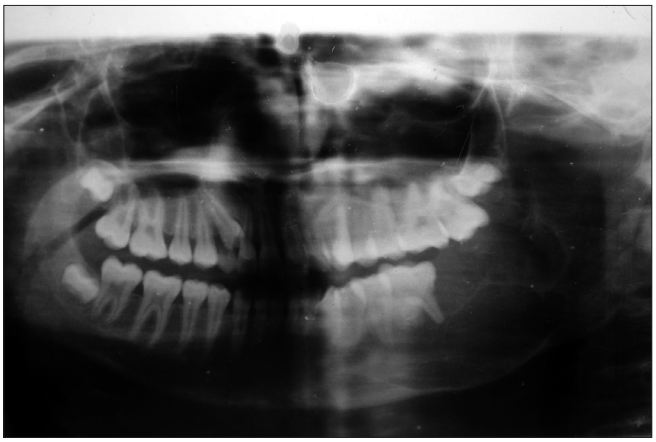

Figure 3: Panoramic radiographic examination of the recurrent lesion showing a multilocular, soap bubble, radioluceny between the anterior border of the left ramus and left second premolar

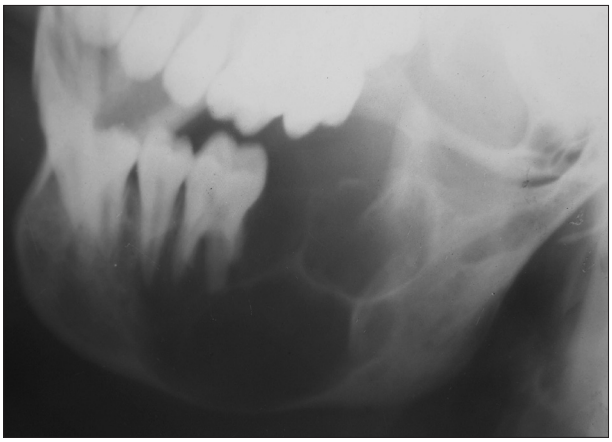

Figure 4: Lateral oblique view radiograph of the recurrent lesion showing a multilocular appearance between the anterior border of the left ramus and left second premolar

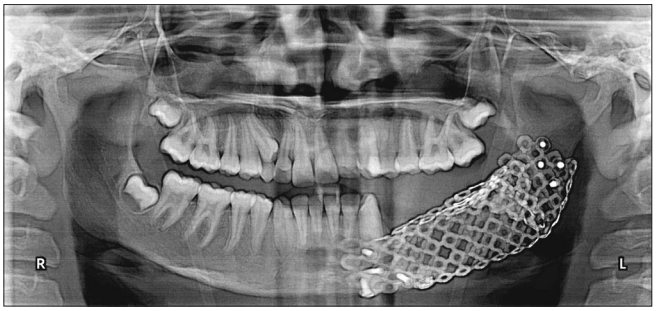

Figure 5: Low cost stainless steel mandibular reconstruction plate fixed immediately after mandibular segmental resection

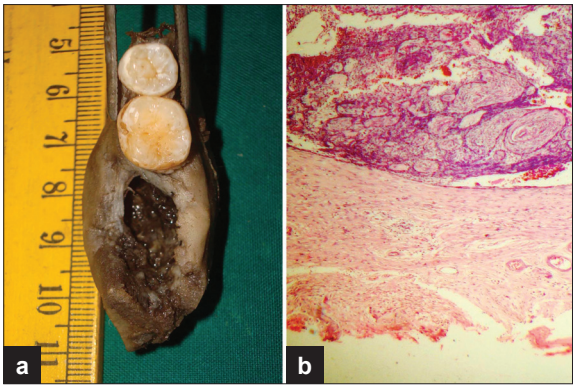

Figure 6: (a) Post surgical radiographic evaluation showing a monocystic lesion with expanded but intact cortical plate and no lower border peroforation. (b) Histopathologic examination of a specimen of the recurrent lesion showing an epithelial lining with intraluminal plexiform (H and E stain, 40×)

[Figures 3 and 4]. Incisional biopsy revealed a cystic lesion consistent with unicystic ameloblastoma. The lesion was treated by segmental resection with immediate reconstruction using a low cost stainless steel mandibular plate [Figure 5]. Post surgical evaluation showed a monocystic lesion with expanded but intact cortical plate. The lower border showed no perforation [Figure 6a]. Microscopically, an epithelial lining with intraluminal plexiform growth was observed [Figure 6b]. Multiple blocks (soft and hard tissues) and serial sections were evaluated; however, neither mural component nor infiltration into bony margins was seen.

DISCUSSION

UCA, strictly, implies not a unilocular radiographic pattern but refers to the monocystic appearance, grossly and microscopically.[2] Radiographically, UCA can present unilocular, pericoronal and multilocular patterns, each with different age predilection.[2] In the current case, a pericoronal radiolucency was found 4 years earlier (primary) and a multilocular radiolucency 4 years later (recurrent). There is no available information in the literature about the dual radiographic pattern between the primary and the recurrent lesion. In Robinson and Martinez, [1] series, the mean age for dentigerous radiographic pattern was 19 years and 47 years for non-dentigerous unilocular lesions. On the other hand, Eversole *et al*,[3] found a mean age of 33 years for multilocular and 19 years for lesions with a unilocular radiographic pattern. These indicate that UCA may show multilocular radiographic pattern in older adults rather than children. In the current case, both the primary and recurrent lesions occurred at a lower age. This is not unusual as others have also found UCA in children less than 15 years of age.[1,4-6] In general, UCA frequently is found in association with mandibular third molars,[2] while it is less frequently found in relation to permanent second molar or premolars.[1,4,5,7].

According to Robinson and Martinez,[1] UCA may show either ameloblastic type of epithelial lining cells or non-ameloblastomatous epithelium. UCA are typed into three subtypes with simple unicystic, luminal and mural categories[8] with implications for difference in biologic behavior.[2,8,9]

There are a number of views regarding the genesis of UCA;[2] de novo origin, from non-neoplastic odontogenic cyst and cystic degeneration of solid ameloblastoma. In our case, both primary and the recurrent lesion displayed ameloblastic epithelium and both were of simple UCA type. A similar finding of identical histology of primary and recurrent lesion was also found by Rosenstein *et al*.[9]

Treatment modalities for UCA include enucleation, enucleation followed by application of Carnoy's solution, marsupialization followed by surgery, and resection.[10] These surgical techniques offer a success rate of 70%, 82%, 84% and 96%, respectively. Several factors are taken in to consideration in the treatment of UCA in children; these include size, location, duration, mural component, psychological impact, control of possible recurrence and scope for follow-up.[4] Psychology plays an important role in the decision making because of its negative impact on growth and function.[4,11] Further, the precise histological subtyping of UCA or distinction from solid ameloblastoma is often difficult to make, prior to definitive surgery.[12] In view of this, primary lesions are treated by means of conservative procedure; however, when odontogenic cell rests or mural component are found in the fibrous wall, it would be prudent to resect uninvolved normal bone along with lesional tissue.[4,8] In our case, odontogenic cell rests were not apparent in the primary lesion and was found only on post surgical evaluation. The short recurrence time in the current case might well be related to the presence of odontogenic rests. The current treatment policy for UCA advocates more aggressive measures, even for primary lesion, such as peripheral osteotomy or segmental resection.[13] In conclusion, a case of UCA in a child with a dual radiographic pattern is presented.

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How to cite this article: Ponniah I. Recurrent unicystic ameloblastoma in a child. J Oral Maxillofac Pathol 2011;15:236-8.

Source of Support: Nil. **Conflict of Interest:** None declared.